

PEDIATRICS®

OFFICIAL JOURNAL OF THE AMERICAN ACADEMY OF PEDIATRICS

Risk Factors Associated With Deformational Plagiocephaly

Jessica L. Joganic, John M. Lynch, Timothy R. Littlefield and Brian C. Verrelli

Pediatrics published online Nov 16, 2009;

DOI: 10.1542/peds.2008-2969

The online version of this article, along with updated information and services, is located on the World Wide Web at:

<http://www.pediatrics.org>

PEDIATRICS is the official journal of the American Academy of Pediatrics. A monthly publication, it has been published continuously since 1948. PEDIATRICS is owned, published, and trademarked by the American Academy of Pediatrics, 141 Northwest Point Boulevard, Elk Grove Village, Illinois, 60007. Copyright © 2009 by the American Academy of Pediatrics. All rights reserved. Print ISSN: 0031-4005. Online ISSN: 1098-4275.

American Academy of Pediatrics

DEDICATED TO THE HEALTH OF ALL CHILDREN™



Risk Factors Associated With Deformational Plagiocephaly

AUTHORS: Jessica L. Joganic, BA,^{a,b,c} John M. Lynch, PhD,^{a,b} Timothy R. Littlefield, MS,^d and Brian C. Verrelli, PhD^{b,c}

^aBarrett Honors College, ^bSchool of Life Sciences, and ^cCenter for Evolutionary Functional Genomics, Biodesign Institute, Arizona State University, Tempe, Arizona; and ^dCranial Technologies, Inc, Phoenix, Arizona

KEY WORDS

child care, multivariable analysis, prenatal exposure, sleep position

ABBREVIATIONS

DP—deformational plagiocephaly
 AAP—American Academy of Pediatrics
 CTI—Cranial Technologies, Inc
 NP—natural population
 BL—supine with head turned left
 BR—supine with head turned right
 LS—left side
 RS—right side
 LBW—low birth weight

www.pediatrics.org/cgi/doi/10.1542/peds.2008-2969

doi:10.1542/peds.2008-2969

Accepted for publication Jun 23, 2009

Address correspondence to Brian C. Verrelli, PhD, Arizona State University, School of Life Sciences, Tempe, AZ 85287-4501. E-mail: brian.verrelli@asu.edu

PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

Copyright © 2009 by the American Academy of Pediatrics

FINANCIAL DISCLOSURE: *The authors have indicated they have no financial relationships relevant to this article to disclose.*



WHAT'S KNOWN ON THIS SUBJECT: Previous research has identified several risk factors contributing to the development of DP, including male sex, intrauterine constraint, congenital muscular torticollis, supine sleep position, and plural births. Also, the condition typically lateralizes to the RS of the occiput.



WHAT THIS STUDY ADDS: We analyzed the largest patient database to date with a statistical scheme that allows conclusions about interactions among biological and environmental factors to be made. This study also recognizes novel risk factors, including dizygosity and LBW.

abstract

OBJECTIVE: This study was designed to statistically evaluate the independent and interacting effects of biological and environmental risk factors that influence lateralization of deformational plagiocephaly (DP) in an attempt to provide future guidance for clinical treatment.

METHODS: A database of >20 000 children treated for DP was examined by using 2- and 3-way factor analyses for categorical frequency data, representing the largest statistical analysis of DP to date. Data on parity, zygosity, intrauterine presentation, birth number and weight, sleep position, lateralization, and sex were collected from parents of children with DP who were treated at Cranial Technologies, Inc, from 1990 to 2007.

RESULTS: As with most DP studies, male patients were significantly overrepresented. Nonetheless, after statistically accounting for sex in our analyses, DP is significantly correlated with primiparity, fewer vertex but more breech and transverse intrauterine presentations, twinning (specifically, dizygosity), and, finally, right-sided lateralization. Additional analyses revealed that several factors correlated with DP, such as intrauterine presentation, sleep position, and lateralization, are not easily explained by an underlying biological factor. Instead, sleep position was the single greatest predictor of lateralization.

CONCLUSION: Although previous studies have argued for both environmental and underlying biological factors associated with DP, we found that lateralization in children with DP could be largely explained by environmental factors such as sleep position. *Pediatrics* 2009;124:e000

Deformational plagiocephaly (DP) is the condition that results from the prenatal or postnatal application of external deformational forces to an infant's occiput. It presents primarily as a unilateral flat spot located on the rear of an infant's skull and secondarily in the skull base and facial skeleton, resulting in frontal bossing, misaligned ears, and asymmetrical orbits.^{1,2} In an effort to reduce the incidence of sudden infant death syndrome, which is associated with prone sleeping, the American Academy of Pediatrics (AAP) recommended that parents place their infants in the supine sleeping position.³ Since the actuation of the Back to Sleep campaign in 1992, the percentage of infants slept prone has decreased whereas the number of patients presenting to pediatricians with DP has dramatically increased. Although it is not certain exactly when and how universally this recommendation was adopted, this increase in DP over time suggests that the supine sleeping position might influence the development of DP.^{4–6}

Risk factors highlighted in previous studies include biological (congenital muscular torticollis,^{7,8} male sex,^{9,10} and prematurity¹¹) and environmental factors (primiparity,¹² plural birth,^{6,11,13} and intrauterine constraint).^{11,14} Although there are certain characteristics that are similar among studies, such as children with DP having predominantly right-side flat spots, several are inconsistent and the interaction among these factors and ultimate cause of DP is controversial. In addition, DP studies often include small sample sizes from localized geographic populations for only a suite of factors. Here, the data set examined allows for the largest statistical analysis both in number of individuals and factors simultaneously examined. Our goal was to discover the relative biological and environmental contribu-

tions to the development of DP as well as evaluate those risk factors that may explain the higher prevalence of right-sided plagiocephaly in this population.

METHODS

Clinical Population

The DP sample is of the patient database of Cranial Technologies, Inc (CTI), a company that fashions individualized orthotic headbands for children with nonsynostotic cranial deformations. CTI is headquartered in Phoenix, Arizona, but there are 18 clinics located across 13 of the United States.¹⁵ Patients are referred to CTI only after they are given a diagnosis by their personal physician. Medical history data are recorded with CTI staff questionnaires from the child's parents before treatment begins. These data constitute children seen at clinics spanning the years 1990–2007. Clinics draw from nationally distributed populations and treat children 6 to 24 months of age. Although ethnicity and socioeconomic status are important factors, these personal and demographic data are currently not available to CTI. Thus, conclusions from this sample are currently limited to physical factors associated with the overall DP population and not to any 1 demographic area.

As of 2007, the CTI database included 31 691 patients. We removed patients who were treated postoperatively after surgery for craniosynostosis or who had other obvious confounding conditions (eg, hydrocephalus, achondroplasia, Down syndrome; $n = 2624$). Because we were evaluating risk factors specifically influencing lateralization (eg, bias of right-sided to left-sided plagiocephaly), we removed patients who had been given a diagnosis of either symmetric dolichocephaly (scaphocephaly) or symmetric brachycephaly ($n = 8376$). This resulted in a final sample size of 20 691

individuals, representing the largest analysis of DP to date.

Factors Measured

Data collected by CTI on sex, parity, zygosity, intrauterine presentation, birth number and birth weight, sleep position, and lateralization were used for this study, and were chosen on the basis of previous research conducted in individuals with DP and on availability of "natural population" (NP) data. Similar to other studies that have compared patient and NP samples,¹⁶ we collected published statistics on population averages from obstetric texts and available annual statistics reports produced for all subpopulations from the United States to compare with our DP sample.^{17–19}

The sex of the child is an obvious candidate factor because it reflects a biological component, as male fetuses are larger and grow more rapidly than female fetuses, thus experiencing a more constrained intrauterine environment.^{6,10,20} In addition, compared with female fetuses, male fetuses are less flexible as a result of decreased estrogen during the perinatal period, which makes them more susceptible to deformational anomalies.^{6,21–23} Intrauterine presentation refers to the portion of the fetal body closest to the birth canal—pelvis, shoulder, or head—during the final trimester and is named accordingly: breech; transverse; and vertex. In the vertex presentation, the fetal head is wedged between the maternal pubic symphysis and lumbosacral spine, which can cause localized occipital flattenings.^{6,10} However, in the breech presentation, the fetal head is compressed by the uterine walls and maternal ribcage.²⁴

Birth number was analyzed as singleton or multiples, the latter of which may be a greater factor simply because of intrauterine overcrowding of the fetuses.^{6,11,13,14} The parity of the

mother was recorded as primiparous if the child with DP was her first child or multiparous if the child with DP was not. The potential for DP may be higher in primiparous mothers because their abdominal muscles have not been stretched by previous pregnancies.^{20,24} Zygosity (monozygotic or dizygotic) was considered because, like sex, it may reflect a biological predisposition to the development of DP, as has been suggested by familial studies.^{4,9} Five different sleep positions were considered that reflected a predominant behavior: supine with head turned left (BL), supine with head turned right (BR), on left side (LS), on right side (RS), and prone. Lateralization was measured as the side of the patient's occiput on which the flat spot occurs, either the right or the left.

Although congenital muscular torticollis is a well-documented risk factor,^{7,8} we were unable to evaluate this variable in our study because of its inconsistent diagnosis in our sample population. Many patients present with limited range of motion, neck tightness or head tilt, but no formal diagnosis of torticollis.⁸ Finally, although our database spans the time period of initial recommendation by the AAP that children be slept in a supine position (1992), we could not easily analyze our database to examine the inferred impact this announcement had on the national progression of DP. A major problem is that a discrete time "cutoff" (eg, before and after the announcement) is complicated given that the details of the AAP recommendation changed significantly over the 10 years after its publication, the rate of adoption of protocol across this time period remains unknown, and no data on how different ethnic and socioeconomic groups responded to the recommendation are currently available to us. It should be noted that this problem is not limited to our database and study specifically.

Although a future analysis temporally subsampling the data may be applicable, we recognize that our analysis of DP factors here is actually statistically conservative given the potentially confounding nature of the gradual and heterogeneous transition from prone to supine positions adopted by parents.

Statistical Analyses

Data for the 8 factors in our sample were cross-tabulated by using the JMP 7.0 program (SAS Institute, Inc, Cary, NC). As previously mentioned, because our data came from CTI questionnaires, in some cases data were not available or supplied by the individuals for all factors. Given the size of the database, it was expected that some factor data sets would have different sample sizes compared with others, and accordingly, sample sizes are not the same across all of our statistical analyses. Nonetheless, it should be noted that these unequal sample sizes simply reflect "no data available" for certain individuals and not a systematic bias in how data were collected or analyzed. Because our data are categorical frequency data, we analyzed the various factors and their relative proportions by using several hypotheses that involve expectations based on the NP as a whole (ie, children without DP) or when appropriate and biologically relevant, compared with previously published DP population studies. We examined correlations among factors by using 2- and 3-way "row-by-column" contingency tables (ie, "A × B × C") and evaluated statistical significance with *G* tests of independence, using a Bonferroni correction for *P* values because of multiple comparisons.²⁵ The *G* test is similar to a traditional χ^2 test of independence in computing deviations of observed from expected values; however, it uses a more accurate approximation of the

log-likelihood ratio, on which the χ^2 distribution is based, when the observed sample size is proportionately larger,²⁵ as is the case with our study. We have several hypotheses involving the relationships among the 8 factors for which data were collected. These statistical analyses enable us to examine the correlation between 2 factors while factoring out a third potentially confounding factor in evaluating how our specific factors interact or contribute independently to DP. For example, we test hypotheses such as how lateralization and sleep position are correlated independent of the sex of the child, a possible contributory biological factor, by examining the relationship of sex to the first 2 factors and then statistically factoring it out as an independent covariate. The Bonferroni corrections were necessary because several factors were examined multiple times among different hypotheses, eg, the correlation of intrauterine presentation with lateralization, sleep position, birth number, and sex. Because these hypotheses all involve at least 1 shared factor, this traditional correction factor (ie, *P* value divided by *k*, where *k* is the number of tests involving a specific factor) is a conservative method to account for the lack of independence among tests that would otherwise inflate statistical significance.

RESULTS

Of the 20 691 individuals in our sample, only 1006 did not have sex reported, which resulted in a total of 13 158 male patients and 6527 female patients; a nearly perfect 2:1 ratio of male patients to female patients. Our DP sample significantly deviates from the ~50:50 ratio in the NP (*G* = 1150; *P* < .00001), indicating a statistically greater number of male patients with DP than female patients with DP. Other published studies of DP are also consistent with a greater number of male patients,^{9,12,26,27} and in fact, like the cur-

TABLE 1 Factors of Patients With DP and Statistical Contrasts With Expected Ratios

Factor	Male Patients, <i>n</i>	Female Patients, <i>n</i>	Total Observed Ratio, %	Expected Ratio, % ^a
Singleton birth weight				
Normal, >2500 g	9379	4361	90.2	93.8 NP ^b
LBW, <2500 g	959	538	9.8	6.2
Parity				
Primiparous	6567	3363	52.4	50 NP ^b
Multiparous	6143	2884	47.6	50
Intrauterine presentation				
Vertex	4208	2079	77.5	96.0 NP ^b
Breech	774	470	15.3	3.5
Transverse	369	212	7.2	0.4
Birth number				
Singleton	11224	5321	85.8	96.7 NP ^b
Multiples	1720	1028	14.2	3.3
Zygoty				
Dizygotic	1044	636	88.4	66.7 NP ^b
Monozygotic	138	83	11.6	33.3
Lateralization				
Right	8839	4408	67.3	75 DP ^b
Left	4319	2119	32.7	25

^a "Factor" observed ratios tested against those expected from either observations in the NP or previously published DP data sets, while independently factoring out sex.

^b *P* < .0001.

rent sample, they typically include 2 to 3 times more male patients with DP. Thus, although our sample is significantly composed of male patients, because this observation is consistent with other DP studies, any findings here that differ from other DP studies cannot be explained by sex-ratio bias alone.

Tables 1 through 9 show the statistical tests, their statistical significance, and all data for each of our factor analyses. As stated previously, not all factor data were available for each of the children with DP, and thus, certain combinations of factors have different sample sizes. For example, "lateralization × sleep position" appears in 3 different ta-

bles because 3 different factors were statistically accounted for independently: sex; intrauterine presentation; and zygoty (Tables 4, 6, and 9).

Table 1 shows the data and analyses for 6 of our factors for which we tested hypotheses regarding their differences from the NP or previous DP studies. All tests represent contrasts after factoring out "sex" in 3-way factor analyses. In comparison with the NP percentage of low birth weight (LBW) singleton children (6.2%), which is any newborn weighing < 2500 g,^{18,28} significantly more DP singletons are born LBW in our sample. Primiparous children with DP were significantly greater than multiparous children in our sample.

TABLE 2 G Tests for Correlations Among Birth Number, Intrauterine Presentation, and Sex

Birth No.	Sex	Intrauterine Presentation, <i>n</i>			Correlations ^a		
		Breech	Transverse	Vertex	Test	<i>G</i>	<i>P</i>
Singleton					Birth × sex	9.5	.023
	Male	565	203	3542			
	Female	334	115	1673	Intrauterine × sex	11.3	.023
Multiples					Birth × intrauterine	257	<.0001 ^b
	Male	194	147	593			
	Female	130	88	348			

^a *G* values shown for the correlations between 2 factors independent of the third.

^b Statistically significant after Bonferroni correction.

When compared with published data for the NP for intrauterine presentations,^{18,19} we found significantly more DP births in the breech and transverse presentations. Multiples are far less common compared with singletons in the NP,^{11,13,18,19,28,29} yet the number of multiple births in our sample is still significantly greater than expected. Twice as many dizygotic as monozygotic twins are born annually in the NP^{17–19}; however, there are significantly more dizygotic twins in our DP sample; in fact, nearly eightfold more. Although our DP sample is similar to published DP data in that it is significantly composed of children with right-side flat spots, the ratio of right-side to left-side flat spots in our sample is significantly smaller than in studies of much smaller sample sizes.⁹

Table 2 shows the correlation of birth number and intrauterine presentation. We found that birth number is strongly correlated with intrauterine presentation independent of sex. It is possible that the inflated number of multiples may explain the greater number of nonvertex intrauterine presentations for children with DP, both of which were found to be significantly greater than in the NP (Table 1). Tables 3 to 6 present all correlations among intrauterine presentation, sleep position, lateralization, and sex. As the fetus becomes constrained in utero, pressure from the maternal pelvis could cause DP, and thus, intrauterine presentation could determine lateralization. Pressure from predominantly lying on 1 side of the head ("sleep position") in early postnatal development could have a similar effect. In addition, it has been proposed that intrauterine presentation may predict sleep position.³⁰ Our results show that intrauterine presentation and sleep position are not correlated when factoring out sex and lateralization (Tables 3 and 6), nor are lateralization and intrauterine

TABLE 3 *G* Tests for Correlations Among Intrauterine Presentation, Sleep Position, and Sex

Intrauterine Presentation	Sex	Sleep Position, <i>n</i>					Correlations ^a		
		BL	BR	LS	RS	Prone	Test	<i>G</i>	<i>P</i>
Breech	Male	177	419	4	18	9	Intrauterine × sex	20.3	.027
	Female	113	246	1	17	10			
Transverse	Male	102	190	6	7	4	Sleep × sex	14.7	.261
	Female	48	119	1	4	2			
Vertex	Male	1141	2307	25	74	27	Intrauterine × sleep	26.1	.052
	Female	559	1091	18	37	24			

^a *G* values shown for the correlations between 2 factors independent of the third.

TABLE 4 *G* Tests for Correlations Among Lateralization, Sleep Position, and Sex

Lateralization	Sex	Sleep Position, <i>n</i>					Correlations ^a		
		BL	BR	LS	RS	Prone	Test	<i>G</i>	<i>P</i>
Left	Male	3190	156	69	26	39	Lateralization × sex	3.2	.669
	Female	1505	62	47	11	22			
Right	Male	138	6615	23	280	68	Lateralization × sleep	15378	<.0001 ^b
	Female	75	3245	14	127	44			

^a *G* values shown for the correlations between 2 factors independent of the third.

^b Statistically significant after Bonferroni correction.

TABLE 5 *G* Tests for Correlations Among Lateralization, Intrauterine Presentation, and Sex

Lateralization	Sex	Intrauterine Presentation, <i>n</i>			Correlations ^a		
		Breech	Transverse	Vertex	Test	<i>G</i>	<i>P</i>
Left	Male	226	123	1404	Lateralization × sex	1.4	.696
	Female	148	64	701			
Right	Male	548	246	2804	Lateralization × intrauterine	7.1	.133
	Female	322	148	1378			

^a *G* values shown for the correlations between 2 factors independent of the third.

TABLE 6 *G* Tests for Correlations Among Intrauterine Presentation, Sleep Position, and Lateralization

Intrauterine Presentation	Lateralization	Sleep Position, <i>n</i>					Correlations ^a		
		BL	BR	LS	RS	Prone	Test	<i>G</i>	<i>P</i>
Breech	Left	280	16	4	5	8	Intrauterine × lateralization	5.2	.879
	Right	21	671	1	30	11			
Transverse	Left	150	5	4	1	2	Intrauterine × sleep	18.6	.289
	Right	9	322	3	10	5			
Vertex	Left	1702	80	31	10	19	Lateralization × sleep	6838	<.0001 ^b
	Right	88	3523	18	105	38			

^a *G* values shown for the correlations between 2 factors independent of the third.

^b Statistically significant after Bonferroni correction.

presentation correlated after factoring out sex and sleep position (Tables 5 and 6). After factoring out sex and in-

trauterine presentation, sleep position and lateralization are significantly correlated (Tables 4 and 6). Specifically,

children with DP who sleep on their RS and LS tend to have right-side and left-side flat spots, respectively.

Tables 7 to 9 present correlations among zygoty, lateralization, sleep position, and sex. Examining differences between monozygotic and dizygotic twins can reveal whether a biological component (other than sex) may explain lateralization and sleep position for children with DP. We find no independent correlation between zygoty and either lateralization or sleep position after factoring out the other, respectively (Tables 7 and 8), yet lateralization and sleep position are correlated when factoring out zygoty (Table 9), which is true independent of sex (Table 4).

DISCUSSION

As with other studies, our DP sample is significantly composed of male patients.^{6,9,11,13,20,24,27} The degree of skeletal maturity, a measure of the extent to which bones have achieved adult shape and configuration, is advanced in females at all ages from infancy through adolescence.³¹ Whether this translates into reduced skeletal strength of the infant cranium in male patients with DP is unclear but several factors, including timing of development, sex-chromosome effects, and hormone levels, have been suggested.^{32–34} Others have suggested that male infants are heavier than female infants and may thus exert an increased force on their fragile and plastic cranial bones when lying in a supine position.^{9,12,26,27} However, although our male patients with DP are, on average, heavier than our female patients with DP, both sexes are significantly lighter than the average NP birth weight of 3317 g.²⁸

It is not surprising that our sample of children with DP, as well as those studied by others,^{11,13,20} would be associated with a greater number of multi-

TABLE 7 G Tests for Correlations Among Zygosity, Lateralization, and Sex

Zygosity	Sex	Lateralization, <i>n</i>		Correlations ^a		
		Left	Right	Test	<i>G</i>	<i>P</i>
Dizygotic	Male	362	682	Zygosity × sex	2.8	.247
	Female	200	436	Lateralization × sex	3.5	.172
Monozygotic	Male	40	98	Zygosity × lateralization	2.9	.230
	Female	31	52			

^a *G* values shown for the correlations between 2 factors independent of the third.

TABLE 8 G Tests for Correlations Among Zygosity, Sleep Position, and Sex

Zygosity	Sex	Sleep Position, <i>n</i>					Correlations ^a		
		BL	BR	LS	RS	Prone	Test	<i>G</i>	<i>P</i>
Dizygotic	Male	299	503	4	12	10	Zygosity × sex	8.2	.145
	Female	147	322	4	10	7	Sleep × sex	11.4	.180
Monozygotic	Male	32	62	1	4	3	Zygosity × sleep	10.4	.238
	Female	25	41	0	0	2			

^a *G* values shown for the correlations between 2 factors independent of the third.

TABLE 9 G Tests for Correlations Among Zygosity, Sleep Position, and Lateralization

Zygosity	Lateralization	Sleep Position, <i>n</i>					Correlations ^a		
		BL	BR	LS	RS	Prone	Test	<i>G</i>	<i>P</i>
Dizygotic	Left	439	18	3	0	6	Zygosity × lateralization	4.9	.426
	Right	23	851	5	22	14	Zygosity × sleep	8.8	.359
Monozygotic	Left	58	2	1	1	1	Lateralization × sleep	1554	<.0001 ^b
	Right	2	104	1	3	5			

^a *G* values shown for the correlations between 2 factors independent of the third.

^b Statistically significant after Bonferroni correction.

ple, rather than singleton, births compared with the NP. In fact, multiples are more likely to be born premature because of increased uterine distention,^{18,19} and prematurity has obvious implications for reduced motor coordination and activity levels that can be prime conditions for the development of DP. As such, prematurity data should be included in future studies of children with DP.

Previous studies have shown a higher incidence of DP within families, suggesting either a possible genetic component or repetitive familial child care practices.^{9,13} Comparisons of dizygotic and monozygotic twins are typically used to reflect genetic versus environmental contributions, yet our analysis

shows no difference between these 2 twinning types for DP. Thus, our observations would suggest that, at face value, DP may have no underlying genetic component, yet this does not necessarily mean that certain aspects of DP, such as neonatal postural preferences,^{35,36} are not influenced genetically. Nonetheless, these data may provide support for shared or repetitive child care practices within families. Thus, this explanation, and not a genetic one, may potentially explain the previously documented higher frequency of flat spots within families. It is possible that our sample is genetically different from other DP samples yet, given that our sample is from a large geographic patient base, this is

unlikely. Thus, it is more likely that a number of genetic loci, all with small individual effects on average, may differ across ethnic groups, populations, and even families, such that no single gene is solely responsible for DP. To date, this is the first study of this magnitude to examine the role of zygosity in DP and one of the few addressing plurality at all.^{11,13,14,20} Although we find a general lack of an underlying genetic correlation with DP, of interest is the significantly greater ratio of dizygotic to monozygotic twins with DP. Given the well-documented, recent international increase in the use of fertility-enhancing drugs, which have been shown to retard fetal growth, lower birth weight, and produce fraternal twins,^{17,18,37} it is possible that this explains the inflated number of dizygotic twins in our sample and is a major contributor to DP.

Although a subject of much debate, postural asymmetries are noted in neonatal infants with a preference for spontaneously turning the head to the right when lying supine.^{37,38} Studies noting that this head-turning preference is not displayed by premature infants in the weeks after birth suggest that factors both inherent to the fetus and within the intrauterine environment in the final weeks of pregnancy influence this supine postural preference.^{30,38} We found that intrauterine presentations do not explain the recognized trends in sleep position and lateralization of DP. In fact, the increase in DP nonvertex presentations may be because of the greater number of multiple birth infants, because multiples frequently present with at least 1 fetus breech or transverse as a result of increased intrauterine constraint, which is supported by the correlation between birth number and intrauterine presentation found here. Finally, our finding that primiparity and DP are correlated is in accordance

with previous work and suggests that the maternal abdominal muscles may constrain the intrauterine environment and restrict full range of fetal motion.^{7,11,15} Nonetheless, our analyses reveal complex relationships associated with intrauterine presentation and raise additional questions. For example, intrauterine position (eg, left occiput anterior or right sacral posterior), may be a better predictor of the location of flat spots that could then develop into DP in a preferred sleep position, particularly because the twin carried in the lower intrauterine position is more likely to develop DP than the twin carried in the higher intrauterine position.¹³

Finally, although some have found no correlation between sleep position and lateralization,²⁰ others have suggested that sleep position can exacerbate a preexisting localized flat spot caused by either the intrauterine envi-

ronment or passage through the birth canal because the infant's head will rest on the flat spot by force of gravity and lack of motor control.^{4,5,39,40} Interestingly, after controlling for several environmental and biological factors, including intrauterine presentation, sleep position is the 1 environmental factor that consistently and independently explains lateralization in our study. On a related note, past studies have identified torticollis as an important factor,^{7,8} thus, given that we show sleep position and, specifically, head positioning are linked to DP, future studies must be designed a priori to examine torticollis, independent of and in conjunction with sleep position.

CONCLUSIONS

When interactions among factors are examined together, DP risk seems to be associated with males, firstborns, LBW, breech or transverse intrauterine presentation, and multiple birth in-

ants, specifically dizygotic twins. However, independent of these biological and environmental factors, it seems that sleep position is the best predictor of DP. Thus, future research must be designed a priori to resolve the interactions among environmental factors for effective prevention, rather than simply treatment. Examination of novel factors, such as antenatal practices, prenatal health care, and the maternal environment within and between closely related children, may elucidate the controversial and poorly understood etiology of DP.

ACKNOWLEDGMENTS

We greatly appreciate the provisioning of data by CTI, the access provided to the computing resources of Mark Spencer, PhD, the initial statistical input provided by Gary Schwartz, PhD, comments made by Lynn Copes, and the time and effort of Kristi Lewton, Audrey Martyn, and Terry Ritzman.

REFERENCES

- Besson A, Pellerin P, Doual A. Study of asymmetries of the cranial vault in plagiocephaly. *J Craniofac Surg*. 2002;13(5):664–669
- David DJ, Menard RM. Occipital plagiocephaly. *Br J Plast Surg*. 2000;53(5):367–377
- American Academy of Pediatrics, Task Force on Infant Positioning and SIDS. Positioning and SIDS. *Pediatrics*. 1992;89(6 pt 1):1120–1126
- Hutchison BL, Thompson JMD, Mitchell EA. Determinants of nonsynostotic plagiocephaly: a case-control study. *Pediatrics*. 2003;112(4). Available at: www.pediatrics.org/cgi/content/full/112/4/e316
- Kane AA, Mitchell LE, Craven KP, Marsh JL. Observations on a recent increase in plagiocephaly without synostosis. *Pediatrics*. 1996;97(6 pt 1):877–885
- Peitsch WK, Keefer CH, LaBrie RA, Mulliken JB. Incidence of cranial asymmetry in healthy newborns. *Pediatrics*. 2002;110(6). Available at: www.pediatrics.org/cgi/content/full/110/6/e72
- Clarren SK. Plagiocephaly and torticollis: etiology, natural history, and helmet treatment. *J Pediatr*. 1981;98(1):92–95
- Golden KA, Beals SP, Littlefield TR, Pomatto JK. Sternocleidomastoid imbalance versus congenital muscular torticollis: their relationship to positional plagiocephaly. *Cleft Palate Craniofac J*. 1999;36(3):256–261
- Chaddock WM, Kast J, Donahue DJ. The enigma of lambdoid positional molding. *Pediatr Neurosurg*. 1997;26(6):304–311
- Mulliken JB, Vander Woude DL, Hansen M, LaBrie RA, Scott RM. Analysis of posterior plagiocephaly: deformational versus synostotic. *Plast Reconstr Surg*. 1999;103(2):371–380
- Littlefield TR, Kelly KM, Pomatto JK, Beals SP. Multiple-birth infants at higher risk for development of deformational plagiocephaly. *Pediatrics*. 1999;103(3):565–569
- Hutchison BL, Hutchison LAD, Thompson JMD, Mitchell EA. Plagiocephaly and brachycephaly in the first two years of life: a prospective cohort study. *Pediatrics*. 2004;114(4):970–980
- Littlefield TR, Kelly KM, Pomatto JK, Beals SP. Multiple-birth infants at higher risk for development of deformational plagiocephaly: II. Is one twin at greater risk? *Pediatrics*. 2002;109(1):19–25
- Dias MS, Klein DM. Occipital plagiocephaly: deformation of lambdoid synostosis? *Pediatr Neurosurg*. 1996;24(2):69–73
- Cranial Technologies, Inc. Clinic locations. Available at: www.cranialtech.com/ClinicLocations/index.html. Accessed June 14, 2009
- Tong S, Short RV. Dizygotic twinning as a measure of human fertility. *Hum Reprod*. 1998;13(1):95–98
- Hoyert DL, Mathews TJ, Menacker F, Strobino DM, Guyer B. Annual summary of vital statistics: 2004. *Pediatrics*. 2006;117(1):168–183
- Pritchard JA, MacDonald PC, Gant NF, eds. *Williams Obstetrics*. 17th ed. Norwalk, CT: Appleton-Century-Crofts; 1985:503–524
- Ziegel EE, Cranley MS, eds. *Obstetric Nursing*. 7th ed. New York, NY: Macmillan Publishing; 1978:243–256
- van Vlimmermen LA, van der Graaf Y, Boere-Boonekamp MM, L'Hoir MP, Helden PJM, Engelbert RHH. Risk factors for deformational plagiocephaly at birth and at 7 weeks of age: a prospective cohort study. *Pediatrics*. 2007;119(2). Available at: www.pediatrics.org/cgi/content/full/119/2/e408
- Graham Jr JM, ed. *Smith's Recognizable Patterns of Human Deformation*. 3rd ed. Philadelphia, PA: Saunders-Elsevier; 2007
- Liu SH, Al-Shaikh RA, Panossian V, Finerman

- GAM, Lane JM. Estrogen affects the cellular metabolism of the anterior cruciate ligament: a potential explanation for female athletic injury. *Am J Sports Med.* 1997;25(5):704–709
23. Calguneri M, Bird HA, Wright V. Changes in joint laxity occurring during pregnancy. *Ann Rheum Dis.* 1982;41(2):126–128
24. Boere-Boonekamp MM, van der Linden-Kuiper LT. Positional preference: prevalence in infants and follow-up after two years. *Pediatrics.* 2001;107(2):339–343
25. Sokal RR, Rohlf FJ. *Biometry: The Principles and Practice of Statistics in Biological Research.* 3rd ed. New York, NY: Freeman; 1994
26. Bridges SJ, Chambers TL, Pople IK, Wall SA. Plagiocephaly and head binding. *Arch Dis Child.* 2002;86(3):144–145
27. Dias MS, Klein DM, Backstrom JW. Occipital plagiocephaly: deformation or lambdoid synostosis? *Pediatr Neurosurg.* 1996;24(2):61–68
28. Hamilton BE, Miniño AM, Martin JA, Kochanek KD, Strobino DM, Guyer B. Annual summary of vital statistics: 2005. *Pediatrics.* 2007;119(2):345–360
29. Tong S, Caddy D, Short RV. Use of dizygotic to monozygotic twinning ratio as a measure of fertility. *Lancet.* 1997;349(9055):843–845
30. Michel GF, Goodwin R. Intrauterine birth position predicts newborn supine head position preferences. *Infant Behav Develop.* 1979;2:29–38
31. Tanner JM. Physical growth. In: Mussen PH, ed. *Carmichael's Manual of Child Psychology.* New York, NY: John Wiley and Sons; 1970:77–155
32. McPherson GK, Kriewall TJ. The elastic modulus of fetal cranial bone: a first step towards an understanding of the biomechanics of fetal head molding. *J Biomech.* 1980;13(1):9–16
33. Archer J. Sex differences in maturation. In: Connolly KJ, Prechtl HFR, eds. *Maturation and Development: Biological and Physiological Perspectives.* London, United Kingdom: William Heinemann Medical Books; 1981:19–31
34. Hofbauer LC, Khosla S. Androgen effects on bone metabolism: recent progress and controversies. *Eur J Endocrinol.* 1999;140(4):271–286
35. Barnes CL, Cornwell KS, Fitzgerald HE, Harris LJ. Spontaneous head positions in infants during the first 9 postnatal months. *Infant Ment Health J.* 2006;6(3):117–125
36. Previc FH. A general theory concerning the prenatal origins of cerebral lateralization in humans. *Psychol Rev.* 1991;98(3):299–334
37. Ertzeid G, Storeng R, Lyberg T. Treatment with gonadotropins impaired implantation and fetal development in mice. *J Assist Reprod Genet.* 1993;10(4):286–291
38. Gardner J, Lewkowicz D, Turkewitz G. Development of postural asymmetry in premature human infants. *Dev Psychobiol.* 1977;10(5):471–480
39. Moss SD. Nonsurgical, nonorthotic treatment of occipital plagiocephaly: what is the natural history of the misshapen neonatal head? *J Neurosurg.* 1997;87(5):667–670
40. Pollack IF, Losken HW, Fasick P. Diagnosis and management of posterior plagiocephaly. *Pediatrics.* 1997;99(2):180–185

Risk Factors Associated With Deformational Plagiocephaly

Jessica L. Joganic, John M. Lynch, Timothy R. Littlefield and Brian C. Verrelli

Pediatrics published online Nov 16, 2009;

DOI: 10.1542/peds.2008-2969

Updated Information & Services

including high-resolution figures, can be found at:
<http://www.pediatrics.org>

Permissions & Licensing

Information about reproducing this article in parts (figures,
tables) or in its entirety can be found online at:
<http://www.pediatrics.org/misc/Permissions.shtml>

Reprints

Information about ordering reprints can be found online:
<http://www.pediatrics.org/misc/reprints.shtml>

American Academy of Pediatrics

DEDICATED TO THE HEALTH OF ALL CHILDREN™

